Genital Crohn's disease in adolescent boys – A case series

Jagannathan Krishnasamy

From Consultant Paediatrician, Department of Paediatrics, G. Kuppuswamy Naidu Memorial Hospital, Coimbatore, Tamil Nadu, India

Correspondence to: Dr. Jagannathan Krishnasamy, Department of Paediatrics, G. Kuppuswamy Naidu Memorial Hospital, Coimbatore, Tamil Nadu - 641 037, India. E-mail: jagannathkrish@yahoo.co.uk

Received - 06 April 2019 Initial Review - 24 April 2019 Accepted - 01 May 2019

ABSTRACT

Crohn's disease is an inflammatory bowel disease and involvement of the genital region is rare in this condition. Herein, we are reporting three cases of genital Crohn's disease in teenage males who were admitted with a chief complaint of swelling in the penis. Recognizing the symptoms and signs, leading to clinical diagnosis and further confirming it by investigations, is a challenge in this condition. Due to limited literature, managing the disease with available evidence becomes crucial. Both medical and surgical treatments play a key role.

Key words: *Crohn's, Extraintestinal, Genital, Pediatric*

Crohn's disease is an inflammatory bowel disease in which extraintestinal manifestations occur in 25–40% of patients. Genital Crohn's disease is one of the extraintestinal manifestations. There are two modes of presentation of genital Crohn's disease in boys [1,2]. The first one is a direct extension from the bowel by transmural fistulization to the genital region and present as fistula, abscess, ulcer, or edema. The second type is metastatic, in which there is a deposition of granulomas and abscesses involving penis and scrotum [3]. The penile and scrotal edema is assumed to be due to lymphatic obstruction in the hypogastric area or at the base of the scrotum and corpora cavernosum. Cutaneous manifestations are often associated with colonic Crohn's disease as against small bowel Crohn's disease [4]. The case series includes three teenage boys who were presented to the hospital with swelling in the penis.

CASE SERIES

Case 1

A 13-year-old teenager was presented with swelling of the penis (Fig. 1), to the surgical team who subsequently had circumcision. The biopsy of the foreskin revealed multiple granulomata. There were no bowel symptoms. He had a history of perianal abscess 10 years before admitting to the hospital. On examination, he had superficial oral ulcerations and few skin tags in the perianal area. He had upper gastrointestinal endoscopy and colonoscopy which showed cryptitis in the cecum. Rest of the study was normal. He was commenced on aminosalicylates. He remained asymptomatic and is currently under follow-up.

Case 2

A 14-year-old adolescent male was presented to the surgeons with the symptom of penile and scrotal swelling (Fig. 2). The patient had a family history of Crohn's disease where his grandmother was affected. The patient had circumcision and histopathology of the foreskin showed granulomatous lymphangitis. 2 years later, he developed bowel symptoms and weight loss. The endoscopic examination confirmed the diagnosis of Crohn's disease affecting mainly the ascending colon and transverse colon. His bowel disease was treated with intravenous steroids, followed by oral steroids, infliximab, and azathioprine. His penile Crohn's disease was under remission most likely due to a combination of circumcision and medical management of bowel symptoms.

Case 3

A 16-year-old male adolescent was presented with bowel symptoms and raised inflammatory markers. There was a family history of Crohn's disease. He was diagnosed to have Crohn's disease following endoscopy, for which he was managed with aminosalicylate, azathioprine, and adalimumab. He was also being treated for psoriatic arthropathy. 18 months later, he had phimosis and edema of penis and scrotum. It was confirmed as Crohn's disease by histopathology which got better with topical steroids but only for a brief period. The patient had relapse of genital symptoms, which showed improvement after he had received infliximab in place of adalimumab. Later, the patient developed a problem with urine stream, for which he was referred to the surgical team. He is under follow-up now.

DISCUSSION

The diagnosis of genital Crohn's should be suspected in patients with Crohn's disease when they are presented with edema of the genital region, phimosis, or penile ulcers [5]. Genitourinary complications occur in 5–20% of patients having Crohn's disease [6]. In the pediatric population, the metastatic disease

Figure 1: Penile swelling

Figure 2: Penile and scrotal swelling

most often occurs at the same time or precedes the Crohn's disease [7]. Histopathological examination of metastatic genital lesions reveals non-caseating granulomatous inflammation [5].

In our case series, there were varieties of presentations, even though swelling of the penis was the common denominator. In the first case, the child had genital Crohn's disease without bowel symptoms. The second patient developed genital Crohn's before gastrointestinal symptoms. The third patient developed bowel symptoms followed by the genital symptoms.

In the current case series, in the first patient, circumcision had shown to be effective so far, but it would be too early to come to a conclusion. The genital Crohn's in the second patient had been under remission due to the combination of circumcision and infliximab and azathioprine. In the third patient, the disease initially responded to topical steroids but relapsed with more severity. It was active despite offering aminosalicylate, adalimumab, and methotrexate. It responded when we replaced adalimumab with infliximab.

From this case series, it was indicated that circumcision may prove to be useful. Infliximab was found to be effective when other drugs failed to achieve remission.

In the previous studies, metronidazole and prednisolone had been used in combination to achieve remission [8-10]. Following the achievement of remission by the above regime, different methods can be used to maintain remission. In a case report by Lane *et al*., maintenance of remission was done using regular metronidazole, minimal dose of prednisolone, and azathioprine [9]. Oral steroids (alone) were used for remission by Gomez *et al*. [10] Remission of genital Crohn's disease could also be achieved by topical steroids [11] or intralesional steroid injection [12]. Many previous case reports have shown evidence of the role of infliximab in refractory metastatic genital Crohn's disease [13-15]. Patients who failed to respond to the combination of prednisolone and azathioprine have shown response to the addition of infliximab [13]. There could be a complete resolution of symptoms by the end of infliximab induction [15].

Surgical management of penile Crohn's includes surgical plastic reconstruction or surgical plastic suturing [16], split-thickness skin graft [17], circumcision [10], regular curettage, and radical excision of penile and scrotal skin [18]. Circumcision is found to be a useful surgical option in penile Crohn's disease on its own or in adjunct with medical treatment [19,20]. The treatment has to be tailored down to each patient. Large multicenter trials or meta-analysis of the treatment of genital Crohn's would be useful to formulate guidelines to standardize the treatment.

CONCLUSION

When we encounter genital lesions in patients with Crohn's disease, we should have a high index of suspicion to diagnose genital Crohn's disease. When the genital involvement precedes the gastrointestinal symptoms and if we suspect Crohn's disease, every effort should be made to prove or disprove the diagnosis, including colonoscopy screening. From our study, circumcision and infliximab have been found to be useful in achieving remission, either on its own or in combination.

REFERENCES

1. Minocha A, Anderson MW, Eaton BG, Harty RF. Crohn's disease complicating male genitourinary tract without overlying cutaneous involvement. Am J Gastroenterol 1996;91:1463-4.
2. Feller ER, Ribaudo S. Gynecologic aspects of crohn's disease. Am Fam Physicians 2001;64:1725-8.
3. Ploysangam T, Heubi JE, Eisen D, Balistreri WF, Lucky AW. Cutaneous crohn's disease in children. J Am Acad Dermatol 1997;36:697-704.
4. Arm JP, Mark EJ. Case records of the Massachusetts general hospital. N Engl J Med 2000;343:127-33.
5. Zelhof B, Biyani CS, Anathhanam AJ, Pollock B, Browning AJ. Severe penile edema: An unusual presentation of metastatic crohn disease. Int J Urol 2006;13:189-91.
6. Simoneaux SF, Ball TI, Atkinson GO Jr. Scrotal swelling: Unusual first presentation of crohn's disease. Pediatr Radiol 1995;25:375-6.
7. Palamaras I, El-Jabbour J, Pietropaolo N, Thomson P, Mann S, Robles W, *et al.* Metastatic crohn's disease: A review. J Eur Acad Dermatol Venereol 2008;22:1033-43.
8. Ninan T, Aggett PJ, Smith F, Youngson G, Miller ID. Atypical genital involvement in a child with crohn's disease. J Pediatr Gastroenterol Nutr 1992;15:330-3.
9. Lane VA, Vajda P, King D, Stahlschmidt J, Sugarman I, Subramaniam R, *et al.* Metastatic crohn's disease: Two cases of penile crohn's and literature review. J Pediatr Urol 2010;6:270-3.
10. Gomez JM, Salinas CS, Usabiaga IA. Metastatic crohn's disease in

childhood. An Esp Pediatr 2001;55:165-8.
11. Chiba M, Iizuka M, Horie Y, Masamune O. Metastatic crohn's disease involving the penis. J Gastroenterol 1997;32:817-21.
12. Vint R, Husain E, Hussain F, McClinton S, Ormerod A. Metastatic crohn's disease of the penis: Two cases. Int Urol Nephrol 2012;44:45-9.
13. Escher JC, Stoof TJ, van Deventer SJ, van Furth AM. Successful treatment of metastatic crohn disease with infliximab. J Pediatr Gastroenterol Nutr 2002; 34:420-3.
14. Miller AM, Elliott PR, Fink R, Connell W. Rapid response of severe refractory metastatic crohn's disease to infliximab. J Gastroenterol Hepatol 2001; 16:940-2.
15. Lee Y, Lee YM, Kim MJ, Choe YH. Penile crohn's disease resolved by infliximab. J Crohns Colitis 2013;7:e146-9.
16. Corazzaa M, Ughib G, Spisanib L, Virgilia A. Metastatic ulcerative penile crohn's disease. J Euro Acad Dermatol Venereol 1999;13:224-6.
17. Black PC, Friedrich JB, Engrav LH, Wessells H. Meshed unexpanded split-thickness skin grafting for reconstruction of penile skin loss. J Urol 2004; 172:976-9.
18. Cockburn AG, Krolikowski J, Balogh K, Roth RA. Crohn disease of penile and scrotal skin. Urology 1980;15:596-8.
19. Sackett DD, Meshekow JS, Figueroa TE, Napoli JA. Isolated penile lymphedema in an adolescent male: A case of metastatic crohn's disease. J Pediatr Urol 2012;8:e55-8.
20. Rodriguez RM, Salido EA, Plaza DM, Resina RG, Blasco JM, Martinez PC. Metastatic crohn's disease involving the penile foreskin. Archivos Esp Urologia 2006;59/9:902-5.

Funding: None; Conflict of Interest: None Stated.

How to cite this article: Krishnasamy J. Genital Crohn's disease in adolescent boys – A case series. Indian J Child Health. 2019; 6(5):246-248.

Doi: 10.32677/IJCH.2019.v06.i05.013